to touch over the inner aspects of the thighs. A large, tender deep-lying, nonfluctuant mass was palpated in the left anterior axillary line. The bladder was greatly distended; 1250 cc. of urine was removed by catheter.

The important laboratory findings were the following: Leukocytes numbered 14,600 per cu. mm. of blood—neutrophils 84 per cent, small lymphocytes 8.5 per cent, large lymphocytes 2.5 per cent, monocytes 4 per cent and eosinophils 1 per cent. The hemoglobin content was 10.2 gm. per 100 cc. and erythrocytes numbered 3,850,000 per cu. mm.

The spinal fluid was xanthochromic and it contained 51 cells per cu. mm., 85 per cent of them lymphocytes and 15 per cent neutrophils. The protein content was 525 mg. per 100 cc. The pressure of fluid was 22 cm. of water; it did not increase upon jugular compression. Pantopaque® was introduced and a complete block at the level of the first lumbar vertebra was noted radiographically.

The patient was given 600,000 units of procaine penicillin G, 200,000 units of buffered crystalline sodium penicillin G, 0.50 gm. of streptomycin and 0.50 gm. of dihydrostreptomycin intramuscularly and was taken to the operating room four hours after admittance. An incision was made exposing the spines and lamina of the twelfth thoracic and the first lumbar vertebrae. The subcutaneous tissues were edematous and there was little bleeding. Upon removal of the lamina exposed, a large reddish mass was noted. It was ruptured in an attempt to expose it further and it was observed to be an abscess containing approximately 10 cc. of greenish yellow pus. Cultures were made of the pus. After the area about the lesion was aspirated, it was washed with saline and penicillin solution. The dura appeared normal, and the cord seemed decompressed by the procedure. Two gauze packs impregnated with bismuth tribromphenate were placed in the extradural space, and the skin was closed with interrupted wire retention sutures. Hemolytic staphylococcus grew on the

The postoperative course was uneventful. Catheterization of the bladder was necessary three times. On the eighth postoperative day, the axillary mass was fluctuant and it was incised and drained of 45 cc. of seropurulent material. The patient stayed 11 days in hospital and during that time received 600,000 units of aqueous solution of procaine penicillin and 0.5 gm. of streptomycin every 12 hours, given intramuscularly. At the time of discharge, she had 75 cc. of residual urine after voiding. Upon neurological examination two months after operation, no abnormalities were noted. There was no residual urine.

### SUMMARY

A case of epidural abscess is reported. The condition is rare, but if it is kept in mind in all cases of severe pain in the back and meningeal irritation, and then a history of previous infection is obtained, the diagnosis can almost always be made by infor-

mation supplied by spinal fluid examination. If operation is done early enough in the course of the disease, prognosis is good.

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# "Stiff-Man" Syndrome—Progressive Fluctuating Muscular Rigidity and Spasm

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A PATIENT with symptoms and signs not identifiable with any disease known to the author and, perhaps, not previously described, was observed. A survey of pertinent literature gave no helpful clues. While the patient was being studied, Moersch and Woltman<sup>4</sup> described a small series of 14 similar cases observed over some 30 years at the Mayo Clinic and reported for the first time.<sup>4</sup> They knew of no other such patients and could not fit their findings into an accepted diagnosis.

The following case is presented to call further attention to the syndrome and its possible treatment.

### REPORT OF A CASE

The patient, a 36-year-old white, married citrus rancher, sought medical advice because of "trouble with my legs and arms." It was difficult for him to date the onset of illness. He believed that he had had recurring aching pains and stiffness in his back and shoulders over the preceding 10 years, the episodes growing more frequent and severe. During the previous year the symptoms had spread to the arms and legs and a definite stiffness and jerkiness of movement had been noticed. More recently he had begun to notice impairment of walking and, less frequently, impairment of ability to carry objects. He had become aware that although his strength seemed unimpaired he could not trust the control of his muscles. He illustrated by saying that he could easily lift and carry an 80-pound bag of fertilizer but, quite inexplicably and with no sense of tiring, he would suddenly fall in complete collapse after carrying it

From Cottage Hospital, Santa Barbara. Submitted May 7, 1957. some 15 or 20 feet. Then a moment later he would stand, rise, lift the burden and walk on. He told of similar but less frequent episodes of unexpectedly dropping either light or heavy objects he was carrying in his arms. Moderate aching pains and stiffness were nearly always present diffusely through arms, legs, upper back and the back of the neck and shoulders. The aching, spotty and irregular, became more severe and gradually actual stiffness became more pronounced. Except for the episodes mentioned, he felt that his strength remained unimpaired—if the stiffness did not interfere. He did not consider himself ill.

Seven years before, he had had appendectomy for uncomplicated acute appendicitis. During World War II he had served in Hawaii, the Philippines, Guadalcanal and New Guinea. He had been shot in the right foot but received only an uncomplicated flesh wound. During his service he had also had tonsillitis and, later, dengue fever with complete recovery. He had always worked on ranches and during the preceding ten years had had intermittent exposure, outdoors, to chemical dusts such as DDT and 2-4 D. The dusts were all routinely used commercial products, never to the patient's knowledge causing acute illness.

The family history had no bearing on the illness. Upon physical examination it was observed that the temperature, pulse, respirations and blood pressure were consistently within normal limits. The right upper eyelid drooped very slightly. Cranial nerves were intact. No abnormalities of the pupils and fundi of the eyes were noted. Deep reflexes were normally active and equal bilaterally. Plantar responses were normal. Abdominal and cremasteric reflexes were intact. Sensation was unimpaired. All muscles of the shoulder groups, the entire back and upper and lower legs seemed quite hard and tense. From time to time, on one side or the other, isolated groups of these muscles were noted to be almost rigid—to remain so a few hours or days and then return to the ordinary tense state. No regularity of such changes could be determined.

The result of a test of the blood for syphilis was negative. Erythrocytes numbered 5.5 million per cu. mm. and the hemoglobin content was 16.0 gm. per 100 cc. Mean corpuscular hemoglobin was 29.2 micromicrograms. Leukocytes numbered 11,950 per cu. mm.-43 per cent neutrophils, 2 per cent stabs, 39 per cent lymphocytes, 3 per cent monocytes, 10 per cent eosinophils, and 3 per cent basophils. (Several months later erythrocytes numbered 5.03 million per cu. mm. Hemoglobin content was 15.4 gm. per 100 cc. The hematocrit determination was 39 per cent; mean corpuscular hemoglobin, 31 micromicrograms; mean corpuscular volume, 79 cubic microns; mean corpuscular hemoglobin content, 39 per cent. Leukocytes numbered 13,200 per cu. mm.—neutrophils 49 per cent, stabs 4 per cent, lymphocytes 36 per cent, monocytes 4 per cent, eosinophils 6 per cent, and basophils 1 per cent.)

Results of urinalysis were within normal limits.

No parasites were noted in a specimen of the stool and a guaiac test was negative for blood.

The sedimentation rate (Westergren) was 1 mm. per hour. Agglutination was negative for brucellosis. Nonprotein nitrogen content was 33 mg. per 100 cc. of serum. No lupus ervthematosus cells were found in the blood. Serum potassium content was 5.6 mEq. Fasting blood sugar was 120 mg. per 100 cc.; total protein was 6.1 gm. per 100 cc.—4.1 gm. albumin and 2.0 gm. globulin. Results of skin testing with coccidioidin and histoplasmin were negative; with tuberculin, positive.

The basal metabolic rate was minus 23.

Coproporphyrin in the urine for 24 hours was within a normal range; uroporphyrin and porphobilinogen not present.

No abnormalities were noted in an electrocardiogram.

Results of roentgen examination were: No evidence of pulmonary or pleural disease; heart and mediastinal structures normal in appearance; no evidence of spinal fracture or dislocation or of bone or joint destruction; slight marginal osteoarthritic spur formation in the thoracic spine; numerous small calcifications in the spleen.

No satisfactory diagnosis could be made, and various therapies were tried on an empirical basis. These included administration of salicylates, codeine, sedatives, prednisone, chlorpromazine, physiotherapy, psychotherapy and placebos. None made the slightest difference, and the patient continued to have unexpected falls and to drop objects. After five months, Flexin® (zoxazolamine, 2-amino-5-chlorobenzoxazole) was administered and the dose gradually was raised to 2.0 gm. a day. In the ensuing four months to the time of this report, the patient insisted that his condition was improved; there were no falls or dropping of objects. Upon examination of the muscles, however, no change was noted.

## DISCUSSION

The outstanding features of this case, as in the few others like it that have been reported, were the progressive muscular disability, recurring rigidity of muscles, the patient's falling like a "wooden man" and the fact there was no ascertainable cause for the syndrome. What caused the mild eosinophilia and leukocytosis, conditions not noted in other cases, is not known. Slightly high fasting blood sugar also occurred in one of the other reported cases. In addition, the presence of reducing substances in the urine was observed in four of the reported cases. That and the fluctuating intensity of symptoms led Moersch and Woltman<sup>4</sup> to say, "A metabolic basis for the malady should be considered." The only other abnormality they noted in laboratory studies was a high basal metabolic rate (plus 56, plus 65, plus 77) in three of four patients given this test. "The high rates," these investigators said, "were attributed to muscular activity, since there was no other evidence of hyperthyroidism." In two cases in which muscle biopsy was done, no pathologic changes were observed.

The response to Flexin in the present case was definite, but incomplete. Stiffness, subjectively evaluated, was less and no falls or dropping of objects occurred after administration of the drug was started. Balance in walking, originally very poor, was slightly better but was still poor. Objectively, there was no change in the muscles.

Flexin, a recently introduced mephenesin-like drug chemically unrelated to mephenesin, has been found to be of help in preventing or relieving spasm in voluntary muscles. The site of action is thought to be within the central nervous system upon polysynaptic pathways, where it causes depression with resultant relief of muscular spasm.3 It is not believed to act upon muscle, the myoneural junction, motor nerves or monosynaptic reflex arcs. Use of it has been reported in rheumatic diseases,<sup>5</sup> in cerebral palsy<sup>1</sup> and in a variety of neurological disorders due to disease in either the brain or spinal cord.2 A majority of the patients were appreciably helped. Patients with brain disorders were not helped as much as those with spasticity due to lesions of the upper motor neuron variety. Toxic reactions (chiefly gastrointestinal) generally were minor and all were reversible upon discontinuance of the drug.

# Successful Prochlorperazine Therapy Following Chlorpromazine-Induced Jaundice

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SINCE CHLORPROMAZINE came into general use by physicians in the United States in 1954, chlorpromazine-induced jaundice has been reported in a small number of patients.<sup>1-4</sup> Because of the structural similarity of chlorpromazine and prochlorperazine, and the fact that they are used in similar indications, the present case is of interest in that the patient became jaundiced when treated with chlorpromazine but was subsequently treated with prochlorperazine without complications.

## REPORT OF A CASE

The patient, a 72-year-old white housewife, first observed December 23, 1955, had chronic bronchitis and a moderate anxiety state which was manifested in episodes of dizziness, fatigue, and "nervousness." Chlorpromazine, in daily doses of 25 mg., was administered orally for 11 days. The symptoms were dramatically relieved, and on January 4, 1956, a physical examination showed no significant abnormalities. Results of examination of the blood and urine and determination of serum protein were considered normal, and chlorpromazine therapy was discontinued.

The clinical history of the patient was of some interest. At the age of 17 she had had a stillborn infant at full term. Two spontaneous miscarriages, each

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#### SUMMARY

A case of "stiff-man" syndrome (progressive fluctuating muscular rigidity and spasm) in which treatment with Flexin was moderately helpful is reported.

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after about three months' gestation, followed. At age 26 a hysterectomy was performed for reasons unknown. Hemorrhoidectomy was done at age 57 without complications. Four years before the present illness, the patient had an attack of nausea, vomiting, and diarrhea of several weeks' duration, but this was not accompanied by jaundice and cleared up without specific treatment. She had always considered herself to be extremely nervous.

On January 11, 1956, the patient had an attack of nausea, vomiting, malaise and diarrhea. The temperature was 101° F. Cremomycin®\* and a bland diet were prescribed. In four days the temperature returned to normal.

By January 20 the gastrointestinal symptoms had abated, but in the afternoon of that day the temperature was 99.8° F. The patient had severe pruritus and definite jaundice. She was admitted to hospital in an acutely ill condition. The icteric index was 57 units, thymol turbidity 8 units. The result of a cephalin flocculation test was negative in 48 hours. The cell contents of the blood remained within normal limits. The urine was strongly positive for bile. Symptomatic treatment with fluids, a fat-free diet, Benadryl<sup>®</sup> (diphenhydramine hydrochloride) and mild sedatives was carried out but the patient did not improve.

On January 30 the icteric index was 63 units. Radiographs of the abdomen were considered normal, and a consultant recommended further observation and administration of steroids.

From February 1 to February 8 hydrocortisone was administered, 40 mg. four times daily, and from

<sup>\*</sup>Each 30 cc. contains: "Sulfasuxidine" succinylsulfathiazole, 3.0 gm.; colloidal kaolin, 3.0 gm.; pectin N.F., 0.3 gm.; neomycin sulfate, 300 mg.